

CASE REPORT

A Case Report of Solitary, Intraosseous, Adult-Onset Myofibroma of the Mandible

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Received: 23 June 2010/Accepted: 10 August 2010/Published online: 30 October 2010
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Abstract Myofibroma is a benign proliferation of myofibroblasts in the connective tissue. Solitary intraosseous myofibromas are a rare finding especially in an adult. Myofibroma is a benign lesion and its treatment is similar to any other benign lesion.

This article reports a rare case of a 32 year old lady presenting with a complaint of a painful swelling in the lower third molar region, on examination the tender swelling had expansion of the buccal cortex. The diagnostic work up included OPG radiograph, CT Scan, and a deep incisional biopsy of the mass under L.A. The final diagnosis of myofibroma was made only after the histopathological diagnosis and Immunohistochemistry (IHC). The patient was treated with wide local excision of the lesion in the mandible under GA.

Keywords Mandible · Solitary myofibroma · Intraosseous

Introduction

Myofibromatosis constitute majority of the soft tissue tumors usually seen in the new born. Infantile/congenital myofibromatosis is said to be genetically transmitted. The solitary intraosseous myofibroma is a rare occurrence and literature reports very few cases of intra-osseous myofibroma of mandible.

Case report

A 32 year old lady presented with painful swelling in relation to lower left molar region for a duration 6 months. The swelling gradually increased to the present size. The patient had no habits of tobacco or alcohol abuse and oral hygiene was fair. Patient did not have any systemic illness (non contributory medical history) (Figs. 1, 2, 3 and 4).

OPG radiograph revealed a unilocular well circumscribed homogenous radiolucency extending from the left lower premolar to left lower second molar (Fig. 5). CT scan of mandible showed a well defined expansile osteolytic lesion in the left side of the mandible measuring 4.2 cm by 2 cm with irregular resorption of buccal cortex (Fig. 6).

An incisional biopsy of the mass was done under Local Anesthesia and the specimen was sent for histopathological and immuno-histo-chemical analysis (IHC).

The patient underwent local surgical excision of the mass by peripheral resection of the mandible via an extra oral incision under general anesthesia; the lower margin of the mandible was spared. A stainless steel reconstruction plate was fixed to restore contour. The resected specimen was sent for histopathology and the histopathology report confirmed our previous diagnosis.

The histopathological section was composed of spindle and round shaped cells arranged in diffuse sheets and fascicular pattern; some areas showed interlacing whorled patterns. Spindle and round shaped cells contain plump and ovoid wavy nucleus with eosinophilic cytoplasm. There were dilated slit like vascular spaces with few haemorrhages interspersed in the lesion.

The Immuno-Histo-Chemical analysis revealed biphasic pattern for smooth muscle Actin and Vimentin. Vimentin was strongly positive for slender spindle cells, Actin was strongly positive for plump spindle cells.

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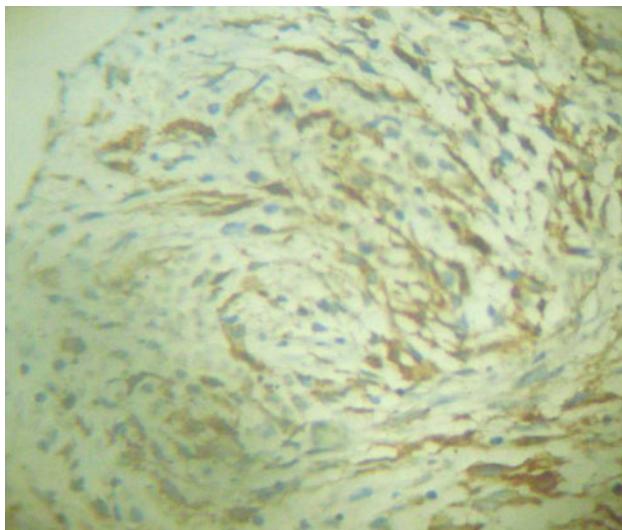


Fig. 1 IHC picture: reactive of antibodies directed against alpha smooth muscle actin

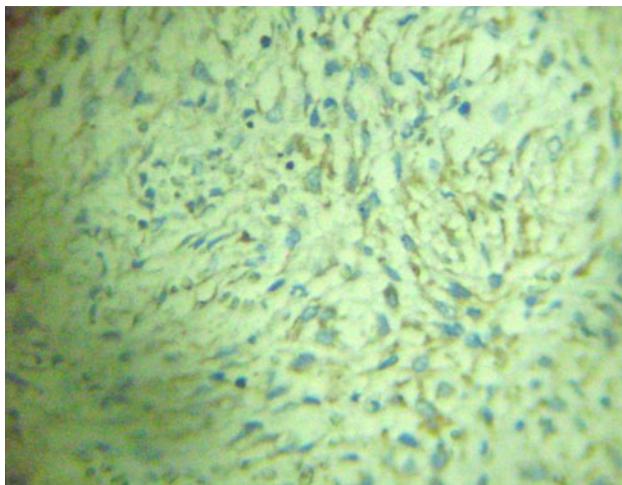


Fig. 2 IHC picture: antibodies immunoreactive against vimentin

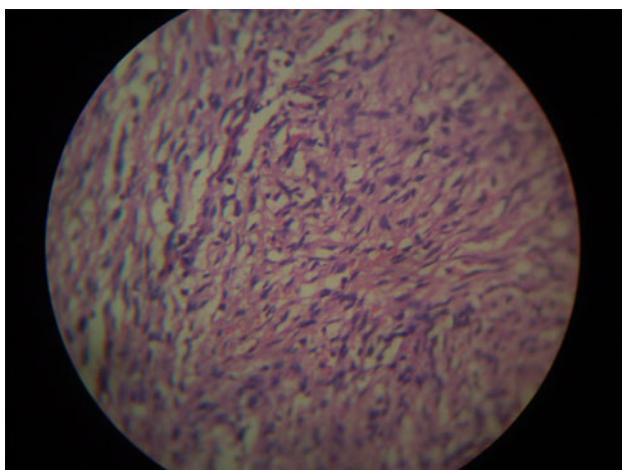


Fig. 3 Spindle and round shaped cells contain plump and ovoid wavy nucleus with eosinophilic cytoplasm. ($\times 40$)



Fig. 4 Extraoral view of patient showing lump in left body of mandible



Fig. 5 OPG showing unilocular homogenous radiolucency in the left body of mandible

Markers CD 68, CD 34, S 100 and Desmin were not reactive. The features found in the specimen were diagnostic of a myofibroma [13] (Figs. 7, 8).

Discussion

The Oral and Maxillofacial region is an uncommon location (2%) for Myofibromas. In the oral cavity, they could be intraosseous, intramuscular or sub mucosal [11]. In the largest series of 79 myofibromas in the oral region, Foss and Ellis reported that approximately one-third of the tumors affected the jaw bones; 12 lesions were central and 15 involved the cortical or periosteal surface but all these

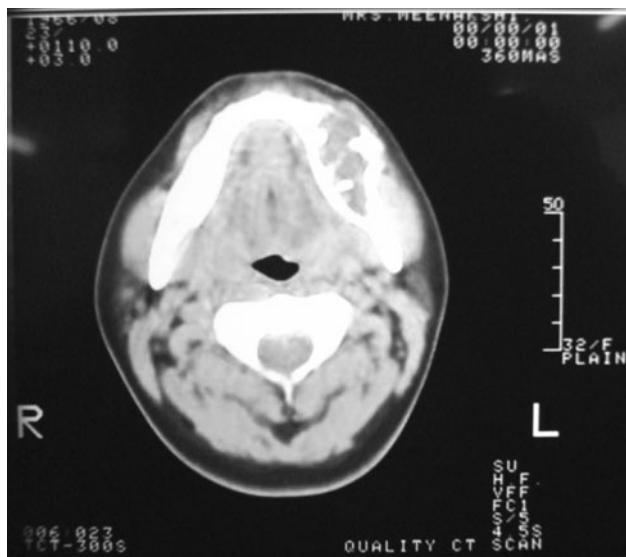
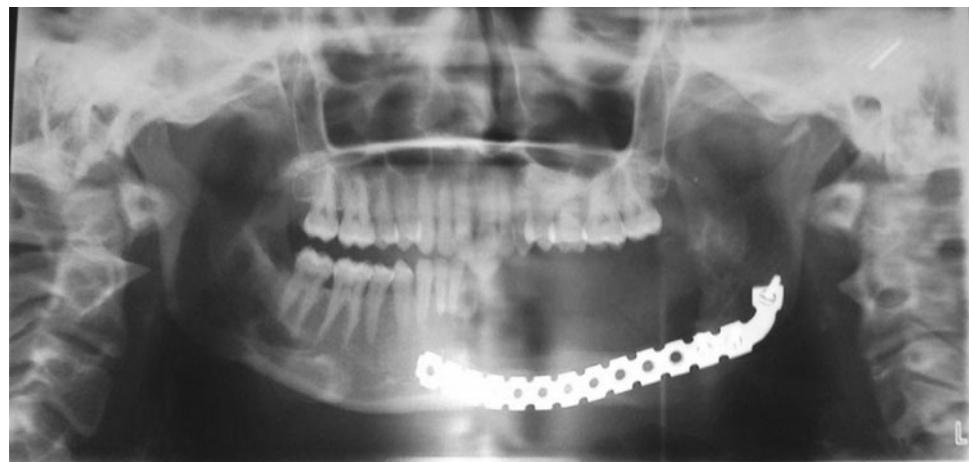


Fig. 6 CT scan shows a well defined expansive osteolytic lesion in the left side of the mandible



Fig. 7 Excised specimen after peripheral osteotomy-lower border spared

Fig. 8 Post operative OPG showing reconstruction plate for contour sparing the lower border



occurred in patients under 18 years of age [7]. Myofibromas have a genetic origin and run in families with dominant inheritance pattern [9]. They should be differentiated from fibromatosis which occurs commonly in the first decade. Fibromatosis occur in the shoulder (22%), chest and back (17%), thigh (13%) and mesentery (10%) [8], they can be present in some uncommon areas in the head and neck like superficial scalp [12], the cervical area causing brachial plexus palsy [14], in the oropharynx compromising airway [6] and even in the orbit [15].

The term Fibromatosis [4] refers to a group of benign soft tissue tumors which have certain characteristics in common, including absence of cytologic and absence of clinical malignant features, a histology consistent with proliferation of well-differentiated fibroblasts, an infiltrative growth pattern, and aggressive clinical behavior with frequent local recurrence.

Myofibromatosis (Multiple) or myofibroma (Solitary) presents with a microscopic appearance similar to that of fibromatosis but the peripheral cells demonstrate eosinophilic cytoplasm remnant of smooth muscle. There is usually a biphasic pattern of lightly staining fibrous areas separated by regions of pericyte-like vascular cell or smooth muscle-like spindle cell proliferations. Histopathological differential diagnosis includes, myofibroma, neurofibroma hamangiopericytoma, nodular fasciitis and leiomyoma.

Immunohistochemistry is done to confirm the diagnosis. In Myofibromas the cells are immunoreactive for vimentin and the smooth muscle actin, but negative or inconsistently positive for desmin or S-100 protein. These stains help to demonstrate the smooth muscle nature of the lesion and to separate myofibromatosis from neurofibroma and leiomyoma, although they are less helpful for nodular fasciitis, which also contains myofibroblasts. Neurofibroma would show positivity for S-100 and Vimeticin, leiomyoma for

desmin and smooth muscle actin, hemangiopericytoma for CD34 along with Vimeticin and smooth muscle actin.

In our case of myofibroma in the mandible (Adult), histological evaluation of the specimen revealed interlacing pattern of spindle-shaped cells with long oval nuclei. Tissue IHC found it to be reactive for antibodies directed against Vimentin and alpha smooth muscle Actin, but not to Desmin [2].

Intraosseous myofibromas present with varied radiological features. In a study of intraosseous myofibromas done by Irit Allon et al. [1] they were found commonly solitary radiolucent lesions located in the mandible of which 70% were unilocular, 30% were multilocular and 67% had well defined borders.

Some myofibromas, especially in the young involute without treatment [10]. Although these tumors are benign, most recurrences are associated with the anatomic restraints during surgery. High recurrences are most likely to be found in cases in which surgical access is difficult. Surgery can range from local excision to partial resection of the mandible. Conservative surgery should be first line of treatment and the patient should be followed up for any recurrent disease [1].

Daimaru et al. [5], Wolfe and Cooper [16] had no recurrences develop in the cases they reported. Alternatively, Chung and Enzinger [3] reported a 10% recurrence rate for the lesions they reviewed. Craig Fowler et al. [8] reports a recurrence rate of 23.8%.

Summary

The Oral and Maxillofacial region is an uncommon location for myofibroma. However, myofibroma should be considered in the differential diagnosis of Unilocular radiolucent lesions especially in children and young adults. Tissue IHC found the cells to be reactive for antibodies directed against Vimentin and alpha smooth muscle Actin.

Treatment of choice is conservative surgery to minimize potential functional and/or aesthetic damage. High recurrences are most likely to be found in cases in which surgical access is difficult.

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